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PREVALENCE OF ANKYLOGLOSSIA IN NEWBORNS OF PALMAS (BRAZIL)

¹Crislla Keroly Xavier Segato, *²Rise Consolação Iuata Costa Rank, ²Joana Estela Rezende Vilela,

¹Luciana Marquez, ¹Tássia Silvana Borges, ³Juliana Batista da Silva
and ²Wataro Nelson Ogawa

¹Department of Dentistry, Lutheran University Center of Palmas - CEULP/ULBRA, Palmas, Tocantins, Brazil
²University of Gurupi - UNIRG, University of Gurupi, Gurupi, Tocantins, Brazil
³Postgraduate, Public health, Northern University of Paraná, Gurupi, Tocantins, Brazil

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*Corresponding author: Rise Consolação Iuata Costa Rank

ABSTRACT

Ankyloglossia is considered a congenital anomaly characterized by the short insertion of the lingual brake. Babies with this anomaly may present restrict tongue mobility, providing swallowing difficulties, deficiency in breastfeeding, and even early weaning. The present study verified the prevalence of ankyloglossia in newborns, frenotomies performed, and the mothers' profile regarding chemical dependence, according to individual medical records from January 2017 to February 2018. This is a documentary analytical cross-sectional research, of a quantitative nature, at the Hospital and Maternity Dona Regina in the capital of Palmas city, State of Tocantins, Brazil. The 5,697 individual medical records with the multidisciplinary evaluation of physicians, dentists and speech therapists and the electronic database of the Joint Accommodation (ALCON) were analyzed. The results showed that Palmas has a 16.1% incidence of ankyloglossia, that males were the most affected (62.5%), and that there was a significant difference for users of illicit drugs, alcohol, and tobacco (p<0.001). Illicit drugs showed a high degree of incidence of this genetic alteration, with 80% of cases. It was concluded that Palmas presents a high prevalence of ankyloglossia when compared to other regions of Brazil and the world, in addition, the effects of maternal chemical addictions seem to be associated with the incidence of the lack of cellular apoptosis of the lingual frenulum of the fetus.

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INTRODUCTION

Ankyloglossia also called a tongue-tie, characterized by non-apoptosis of the sublingual membrane, which depending on the insertion of the frenulum may present restrict tongue mobility (Knox, 2010). This congenitally short lingual frenulum presents as sequelae the deficiency of breastfeeding, difficulty swallowing, lack of adequate breastfeeding causing breast pain, favoring early weaning (Buryk *et al.*, 2010). In Brazil, according to Law n°13,002 June 20, 2014, it is mandatory to carry out the evaluation of the frenulum in newborns (NB) still in the first phase of life, this thanks to the multidisciplinary services offered in hospitals and maternity hospitals (Brazil, 2016). The intraoral examination of the newborn aims to obtain a diagnosis of orofacial functions to observe the tongue

limitations conditions. There is a great variation in the prevalence of ankyloglossia in the world (Forlenza et al., 2010). Joseph et al. (2016), states that there was a 70% increase in the incidence of ankyloglossia between 2004 and 2013 in Canada, and in Spain, the study by Jimenez et al. (2014) reported that the increased prevalence of ankyloglossia was higher than expected. Worldwide, there is no standardization of the protocol for evaluation of the ankyloglossia, but the Bristol Tongue Assessment Tool (BTAT) provides with objectivity, clarity, and simplicity the assessment of the degree of severity of the abnormality lingual frenulum, informing the selection of babies for frenotomy and to monitor the effect of the procedure (Ingram et al., 2019). Frenotomy may be indicated for babies who have difficulties in natural breastfeeding during the first months of life. This surgical technique is efficient to improve posture, tongue mobility and its functions (Coryllos et al., 2004; Ingram et al.,

2015). In Palmas city, the Hospital and Maternity Dona Regina (HMDR) performs intraoral diagnosis of the newborns using the BTAT protocol in children with suspected ankyloglossia. This data began to be filed with HMDR in 2017 in each child's individual medical records. However, no previous statistical data on the prevalence of ankyloglossia in this region of Palmas were found. Thus, the aim of this study was to verify the prevalence of ankyloglossia is still living, frenotomy, and mothers' profile, according to the individual medical records completed by health professionals from January 2017 to February 2018, at the HMDR in Palmas city.

MATERIALS AND METHODS

This is a cross-sectional documentary analytical research, quantitative in the Hospital and Maternity Dona Regina in the state of Tocantins. This study was approved by the Ethics Committee on Human Research PROTOCOL (ECHR): 83146717.0.0000.5516.25. Palmas has a total area of 2219 km2, is located in the geographical coordinates 10° 11' 04" south and 48° 20' 01" west, and parallel 10 South and Meridian 48 West pass through within the territory of the municipality. HMDR de Palmas is considered a reference hospital in the Microregion of Porto Nacional that covers 11 municipalities of the State of Tocantins.

is 9.88 deaths per thousand live births. All 5,697 individual medical records of the Electronic Database of The Joint Accommodation (ALCON) from January 2017 to February 2018 analyzed in this study, there was a multidisciplinary evaluation of physicians, dentists, and speech therapists. Thus, consolidated data on the prevalence of ankyloglossia diagnosis, affecting the type of sex, numbers regarding the intervention of frenotomies of this period and the profile of parents, regarding chemical dependence diagnosed by physicians, were obtained. The data were tabulated in Excel, the analysis of the data performed descriptively with a presentation in the form of tables, in which the clinical findings of the quantity were demonstrated in numbers and percentage of these anomalies. The Chi-square (p<0.05) was used to compare the variables gender of the newborn, frenotomy performed and the presence of maternal chemical dependence (illicit drugs, tobacco, and alcohol).

RESULTS

The HMDR data showed 917 patients born with ankyloglossia (Table 1) the prevalence was 16.1% among the total sample of newborns in the evaluated period. Frenotomy was indicated according to the Bristol protocol and performed in 561 newborns.

Table 1. Distribution in numbers and percentage of ankyloglossia and frenotomy, according to the month and year of the medical records of the Hospital and Maternity Dona Regina in Palmas (Brazil)

Month/year	Ankyl	oglossia	Surger	у	P-value
2017	N	%	N	%	
January	73	19,7	47	12,7	
February	52	14	40	10,8	
March	51	12,4	36	8,8	
April	71	16,7	38	9	
May	72	16,9	37	8,6	
Junho	73	17	47	10,9	
Julho	72	16,2	41	9,25	
August	57	13,4	45	10,58	
September	72	18,7	42	10,90	
October	72	17,7	39	9,60	
November	76	18,9	48	11,57	
December	56	13,1	29	6,82	
2018					
January	58	14,7	40	10,12	
February	62	15,7	32	8,14	0,007*

^{*}Chi-square test with a significance level of p<0.05

Table 2. Distribution in numbers and percentage of participants according to gender, in relation to the presence of ankyloglossia or no, according to individual medical records of the Hospital and Maternity Dona Regina in Palmas (Brazil)

Ankyloglossia	Male		Female		Total children	P-value
	N	%	N	%	N	
Yes	568	19	344	12,6	912	
No	2406	81	2379	87,4	4785	
Total	2974	100	2723	100	5697	0,002*

^{*} \overline{Chi} -square test with a significance level of p < 0.05.

Table 3. Distribution in number and percentage of children with ankyloglossia in relation to chemical-dependent mothers, according to individual medical records of the Hospital and Maternity Dona Regina in Palmas (Brazil)

Ankyloglossia	Drugs		Alcohol		Tabacco		Total		P-value
	n	%	n	%	n	%	n	%	
Yes	8	80	29	21	27	30	64	27,4	
No	2	20	106	79	62	70	170	72,6	
Total	10	100	135	100	89	100	234	100	
									<0,0001*

^{*}Chi-square test with a significance level of p < 0.05.

There is an average of 15 children born per day in this hospital, and according to IBGE data (2017), the mortality rate

The Chi-square statistical test demonstrates that the result among the varieties of treatment was highly significant (p=0.007) for surgical therapy in cases of the presence of an ankyloglossia in the State Reference Hospital. When analyzing the variable gender of children in relation to the presence of ankyloglossia, the result was significant of this genetic alteration to males $(p=0.002; x^2=8.887)$, as 62.3% of male children were found, while girls were affected in 37.7%, the results of gender were practically 2:1 (Table 2). The profile of the parents regarding chemical dependence was highly significant (p<0.0001), in which illicit drugs presented 80% of the cases found, followed by Tabacco with 30% and Alcohol with 21%, in children with the presence of ankyloglossia (Table 3). It was found that illicit drugs present an etiological factor associated with the increase in the presence of ankyloglossia.

DISCUSSION

In the literature, data on the prevalence of ankyloglossia range so far from 1% to 10.7% (Forlenza et al., 2010), with other quantitative studies in which the prevalence in Spain reached 12%, between 2 and 3 times higher than expected, which could be 4% (Jiménez et al., 2014). The growth of incidence of ankyloglossia is also exposed by Joseph et al., (2016), in a study conducted in Canada, in which 459,445 live births obtained 3,022 cases of this change between 2004 and 2013. The study showed that the population incidence of ankyloglossia increased by 70% (rate ratio 1.70, confidence interval of 95%; CI from 1.44-2.01), from 5.0 per 1000 live births in 2004 to 8.4 per 1000 in 2013. However, the present research surpassed these statistical data, sine in Palmas the percentage of 16.1% was found. The profile of the parents regarding chemical dependence was highly significant (p<0.0001), in which illicit drugs presented 80% of the cases found, followed by Tobacco with 30% and Alcohol with 21% in children with the presence of ankyloglossia (Table 3). Depending on the diagnostic criteria used by the authors, the population studied is ankyloglossia, and if the literature data come from observational or series case studies, it will not promote uniformity in diagnostic definition (Forlenza et al., 2010). Thus, this wide variation in the prevalence of ankyloglossia found in the world may be related to the lack of standardization of clinical diagnostic protocols, and also the lack of uniformity in the criteria of evaluation procedures.

Based on scientific evidence and taking into account its potential interference with breastfeeding, it is recommended to early identity ankyloglossia in newborns, also avoiding greater sequelae that will affect the health of the child (Emond et al., 2014). The presence of ankyloglossia is found and if there is a need for surgical intervention, the treatment to be performed consists of a small incision that is performed in the anterior portion of the lingual frenulum, releasing the tongue to perform its movements (Brookes et al., 2014). Regarding surgical treatment at HMDR, after diagnosis with the Bristol protocol, the procedure indicated was the frenotomy technique. This surgery is considered less invasive, with reduced operative time and because it presents satisfactory results for the newborn (Zhu et al., 2017). The high frequency of surgeries performed in newborns in HMDR showed that most cases of ankyloglossia were indicated to receive the surgical procedure. The statistical survey of the present study revealed that the frequency of frenotomy was high in relation to the number of individuals with the short lingual frenulum. However, the Canadian Agency for Drugs and Technologies in Health, (2016) demonstrates uncertainty about the benefits of surgical correction of ankyloglossia. Dixon et al., (2018) stated that when detecting the type of alterations of the lingual frenulum, some may be corrected with myofunctional therapy, and surgery was not required in all cases. In most studies, males appear more affected than females (Suter et al., 2009), according to the present study. Harris et al., (1992) examined in a nursery 500 newborns, ankyloglossia was perceived at a prevalence of 4.4%, was significantly more common in men than in women (6.0% versus 2.3%). Embryological anatomical malformation usually affects men more than women in a 3:1 ratio. With this information, the positive result of males comes from a dominant autosomal disorder, through chromosome X. According to Han et al., (2012), in the dominant character, it is sufficient that one of the parents is a carrier of premutation for the child to receive a copy of the defective gene. In general, boys develop more serious conditions than girls. This happens because they have two X chromosomes, one inherited from their father and another from their mother. If a gene is changed, the normal gene can compensate for the change. Men, however, possess only one X chromosome inherited from their mother and a Y chromosome inherited from their father. If x is damaged, the other is not prepared to meet the deficiency. This type of anomaly happens when the chromosome suffers a break and then a reconstitution. This occurs at random, so an unnatural combination of the chromosome. This explains why the largest number of studies report males as the highest target of an ankyloglossia.

However, according to studies by Baldini et al., (2001) and Vieira (2018), the occurrence of ankyloglossia predominated in females. As ankyloglossia is a congenital condition, it is believed in the influence of the mother's profile on the appearance of this alteration, so data about the parent were considered in the medical records of babies, who could intervene in pregnancy. Of these, the use of illicit drugs, alcohol, and tobacco stood out. According to the World Health Organization (WHO,) (Rea et al., 1999), the term 'drug' is used to 'any substance not produced by the body, which has the property of acting on the system, producing operating changes'. A total of 246 million people - slightly more than 5% of the world's population aged between 15 and 64 years – are estimated to have used illicit drugs in 2013. About 27 million people use drugs, of which almost half are people who use injecting drugs (Beckman, 1984). Teratogens are responsible for developmental disturbances when the exposure of the pregnant woman occurs during the period of the embryo organs are forming. Hereditary and environmental influences can affect embryonic development by altering processes such as intracellular division, cell surface, extracellular matrix, and fetal environment, but they were no hypothesis that confirms this mechanism (World, 2015) The exposure to teratogenic agents may cause damage, such as malformations, changes in physical structures, growth retardation, among others. Its action depends on the stage development dose-effect ratio, maternal-fetal genotype and pathogen-specific mechanism of each agent (Toralles et al., 2009; Schuler et al., 1999). However, research showed that the consumption of chemical dependents can intervene in the involvement of ankyloglossia in the newborn. As evidenced that ankyloglossia is 3.5 times more likely to occur in the drug use series, due to the decrease in mitotic rates (Jimênez et al., 2014). They could found that cocaine use by mothers during pregnancy is a risk factor for ankyloglossia. Mothers' babies with cocaine addiction were three times more likely to develop ankyloglossia than those of healthy mothers (Han et al., 2012). The literature has shown that the index of this genetic alteration seems to be increasing in the world. Thus, further studies related to factors that can promote this high rate of ankyloglossia in this region should be encouraged, since measures for its prevention should be found.

Conclusion: This research contributed significant epidemiological data in relation to ankyloglossia, revealing that the congenital condition is high in this region of Brazil and the data showed more surgical procedures in newborns (frenotomy) in the HMDR. The most affected sex was male 2:1, and maternal dependence on illicit drugs was highly significant findings for the involvement of ankyloglossia in children.

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